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Spontaneous tracheal rupture in a 1-year-old child

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ABSTRACT

Spontaneous tracheal rupture is a rare phenomenon in the pediatric population, however it is a potentially fatal condition; necessitating discussion surrounding controversies of investigation and management. We report a case of an 18-month-old presenting with bilateral pneumothoraces, a pneumomediastinum and dysphagia, with review of the literature. The referring unit inserted one intercostal drain into the left hemithorax despite a chest radiograph demonstrating significant bilateral pneumothoraces. On arrival in our unit the child was in respiratory distress with subcostal recessions, tachypnoea, and bilateral decreased breath sounds. He saturated at 75% on room air and improved to 100% when placed onto polymask oxygen. Subcutaneous emphysema was palpable on both sides of his neck, his chest, and anterior abdominal wall. An intercostal drain was inserted on the right side, improving his respiratory distress. Analgesia and prophylactic antibiotics were prescribed. A contrast swallow was performed to exclude an oesophageal injury, which showed no leak. A Computerized Tomography scan revealed a tear in the membranous trachea posteriorly, from the level of the third thoracic vertebral body extending into the left main bronchus. The patient responded well to conservative management with face-mask oxygen, gentle chest physiotherapy and dietary supplementation. The child never required intubation and did not develop sepsis during his admission. His cough was suppressed with codeine phosphate. On day 10, the chest drains were removed and the child was discharged home 11 days after admission. On follow-up, the child was worked up for complaints of gastro-oesophageal reflux disease. A repeat contrast swallow and oesophageal biopsies confirmed the diagnosis and the child was treated with a laparoscopic Nissan fundoplication and gastrostomy after failure of appropriate medical therapy. No adverse respiratory events have been encountered.

Spontaneous tracheal rupture is a rare phenomenon that occurs most frequently in adults [1]. Diagnosis is often elusive and there is no consensus on treatment strategies. Despite being extremely uncommon in pediatrics this remains an important condition to be aware of, and treatment may be urgent and lifesaving. Tracheal rupture in adults is known to be associated with endotracheal intubation, trauma, chronic steroid usage, acute bronchitis, external beam radiation therapy and acquired tracheomalacia [1–3]. There are a handful of case reports involving spontaneous tracheal rupture in children but to our knowledge this is the youngest case reported.

1. Case report

A 1-year old male presented with a 2-day history of severe coughing and acute onset respiratory distress involving cyanotic episodes, with associated dysphagia. He attended his local hospital in respiratory distress. Primary care included supplemental face-mask oxygen and insertion of a left intercostal drain after a chest radiograph demonstrated a large pneumothorax (Figs. 1 and 2). Despite these measures his clinical condition did not improve and he was referred to our institution with the working diagnosis of esophageal perforation.

On arrival in our unit the child was found to be hypoxic and in significant respiratory distress with tachypnea, subcostal recessions, and decreased breath sounds bilaterally. The intercostal drain was oscillating but not bubbling. Subcutaneous emphysema was palpable on both sides of his neck, down his chest and along his anterior abdominal wall. A second intercostal drain was inserted on the right side and his respiratory distress improved. Chest x-ray

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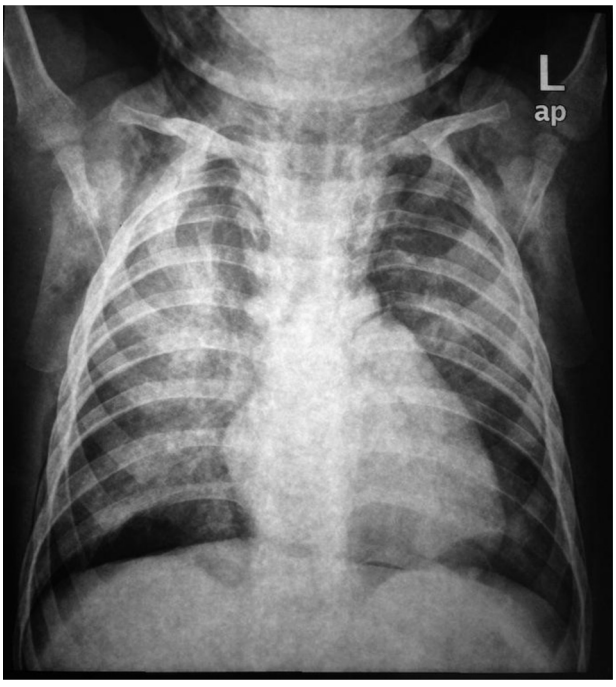

Fig. 1. Chest X-ray with bilateral pneumothoraces and extensive subcutaneous emphysema.

Fig. 3. CT scan with arrow showing tracheal rupture.

revealed smaller bilateral pneumothoraces and a persistent pneumomediastinum with no features of pneumonia.

At this stage the patient was noted to be apyrexial, well hydrated, and not clinically pale, but was documented to be underweight for his age. He was started on analgesia and prophylactic antibiotics, to cover for possible mediastinitis. A contrast swallow was performed, which demonstrated no esophageal leak.

Fig. 2. Lateral neck X-ray.

Computerized Tomography scan (Fig. 3) revealed a longitudinal tear to the posterior membranous tracheal extending from the level of the 3rd thoracic vertebra cranially, into the left main bronchus caudally. After the initial interventions described above, the child stabilized and never required intubation, nor did he develop sepsis during his admission. In the ward he was noted to cough forcefully at night, and when he coughed or cried his chest drains bubbled excessively. Infection was excluded by sending sputum for cultures, and the cough was suppressed with codeine phosphate. The child responded well to conservative management with face-mask oxygen, gentle chest physiotherapy and dietary supplementation. On day 10, the chest drains were removed and he was discharged home 11 days after admission. He was subsequently seen in our outpatient clinic with continued complaints of coughing after feeds. An elective contrast study revealed reflux to the thoracic inlet with no over the top spill and no strictures or features of gastric outlet obstruction. Gastroscopy with biopsy of the distal esophagus and antrum of the stomach was carried out on the following day and the child was started on omeprazole while awaiting biopsy results. Histological examination revealed mild chronic gastritis, and features of mild gastro-oesophageal reflux disease with no cell dysplasia and no Helicobacter Pylori identified. Further questioning revealed a history of multiple chest infections treated as an outpatient and only requiring admission post tracheal rupture. A laparoscopic Nissen fundoplication was performed and a gastrostomy was placed.

2. Discussion

Tracheal injuries are a rare phenomenon in children. Tracheal disruption may occur secondary to direct trauma, which may include blunt or penetrating neck injury, iatrogenic injuries during intubation, or foreign body inhalation. Direct injury is rare in children, as their relatively short neck and mandible protect this vital

structure. In addition, the pediatric trachea is soft, mobile and pliable, further decreasing the incidence of direct traumatic injury. Spontaneous injuries are generally related to raised intra-tracheal pressure against a closed glottis [1]. This is seen in sudden compression injuries, severe coughing, or with associated weakness of the tracheal wall, for example in tracheomalacia secondary to chronic steroid use. The membranous portion of the trachea is the weakest and is exclusively involved in spontaneous tracheal rupture [1]. Patients with spontaneous rupture usually have some preceding history whether this is coughing, vomiting, or choking [2–5]. Acute respiratory distress associated with cervical surgical emphysema is invariably noted.

There is a handful of pediatric cases reported in the English literature. Roh and Lee describe a 7-year-old boy who presented with a 1 cm linear posterior tracheal laceration during an episode of acute tracheobronchitis [2]. The diagnosis was made with plain radiographs, fiber optic endoscopy and CT scans. Their patient was treated conservatively with nasal prong oxygen and antibiotics. They suggest that larger defects require surgical intervention or that the patients should be intubated with the cuff of the endotracheal tube inflated below the level of the laceration and managed in ICU.

Akyol et al. describe a 14-year-old boy with paroxysmal productive coughing, who presented with a posterior tracheal wall rupture [6]. Diagnosis was confirmed with CT scan, tracheobronchoscopy and oesophagoscopy under general anesthesia. The injury was managed by bridging the defect with an endotracheal tube and ventilation in the ICU. The child had an uncomplicated recovery and was well at follow up. The third reported case presented a diagnostic dilemma. Gorosh et al. received a 3 year old boy in their emergency department with a history of progressive swelling and respiratory distress [4]. He was diagnosed with anaphylaxis and treated with cardiopulmonary resuscitation and intubation. Post intubation, bilateral chest drains were placed and the patient's condition improved. Bronchoscopy failed to demonstrate any pathology. The diagnosis was made on a CT scan, which showed a defect in the posterior wall of the trachea, below the level of the endotracheal tube. The patient was repaired by cardiothoracic surgeons and also had a good outcome.

A 14-year-old newly diagnosed diabetic girl is also reported to have ruptured her trachea after days of violent vomiting [5]. She presented with tachypnea and surgical emphysema. A chest x-ray revealed a pneumomediastinum with no pneumothorax. She had a contrast swallow which revealed no oesophageal injury and her tracheal injury was delineated with a CT scan. Bronchoscopy was not deemed necessary. She was also treated with supplemental oxygen and no need for intubation or surgical intervention. Her progression was followed with serial chest x-rays, showing resolution of the surgical emphysema.

Eipe reports a 10-year-old boy who sustained a traumatic laceration of his proximal trachea [7]. This patient was immediately taken to the operating room for bronchoscopy and then insertion of a tracheostomy below the level of the injury. He was intubated blindly and spontaneous ventilation was maintained. Tracheostomy proved to be technically challenging due to the extensive surgical emphysema and the attempt to secure an airway initially failed. A seldinger technique was employed and an airway introduced. No adverse events were subsequently encountered.

This limited experience suggests that even patients with large linear lacerations of the membranous trachea can be treated conservatively without intubation, surgical intervention or ICU admission. Computed tomography is the investigation of choice to delineate the injury without the need for endoscopy, with its potential risks of enlarging the defect and necessitating endotracheal intubation and anesthesia.

3. Conclusion

Spontaneous tracheal rupture presents with an acute massive air leak manifesting as surgical emphysema of the neck and thorax together with respiratory distress. Regardless of these worrying signs most patients can be managed with supplemental oxygen and intercostal drainage of pneumothoraces. Absolute indications for repair include obstruction of the tracheal lumen by mediastinal structures and horizontal lesions extending along more than one third of the tracheal circumference [8]. Ventilation and mediastinitis can also be successfully managed conservatively. Despite the recommendation in most papers advocating bronchoscopy, virtually all reports established the diagnosis and extent of injury on CT scan. Flexible bronchoscopy through the lumen of an endotracheal tube has the risk of missing a covered perforation, whilst rigid bronchoscopy can exacerbate any current tracheal injury. Watchful waiting, without intubation or ICU admission proved to be a safe alternative in our case.

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